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The impact of amelogenesis imperfecta and support needs of adolescents with AI and their parents: an exploratory study.

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Introduction

Amelogenesis Imperfecta (AI) is a rare inherited dental abnormality affecting the structure and clinical appearance of the enamel of the teeth[1]. AI can present as hypoplastic (deficient enamel) or hypomineralised (poorly mineralised enamel), and can be autosomal dominant, recessive and x-linked, as well as some patients having sporadic inheritance patterns. The clinical appearance of AI can be remarkably different between types[2]. The colour of the affected teeth will range from normal to opaque, white or yellow-brown in colour [3]. Teeth are generally weak, easily damaged, and susceptible to decay[4]. Moreover, AI can result in significant tooth disfiguration and may require life-long dental care. The reported prevalence varies considerably with rates ranging from 1:14,000 to 1:700[5].

Psychosocial factors and visible dental defects

The impact of visible disfigurement on psychological health highlights a number of psychosocial challenges including interaction[6] and negative self-perceptions[7]. Welbury & Shaw[8] and Marshman et al[9] reported that children diagnosed with developmental defects of enamel

(which have some similar aesthetic features to AI), experienced considerable teasing. Moreover, recent studies investigating the social judgements made by children in relation to visible damage to the anterior teeth found that negative social judgments may be made on the basis of poor dental appearance, and that aesthetic dental treatment for children may yield important psychosocial benefits[10,11].

Adolescence is a time of particular sensitivity to issues related to appearance and peer relationships, as well as a heightened sense of emotional disturbance [12]. There has been some informative work carried out on the impact of hypodontia (the developmental absence of one or more teeth), within this transition period. A recent in-depth qualitative study[13], found that this condition can have adverse psychosocial effects. One of the key themes from the interviews was the importance of aesthetics. As patients became older they became increasingly more aware of their appearance due to the development of their teeth compared with those of their peers, and that they actively tried to conceal their teeth for aesthetic reasons and reported feeling socially awkward. In contrast, findings from a large-scale quantitative study[14] produced no significant differences between the hypodontia and routine orthodontic groups in terms of psychosocial impact. However, the authors acknowledge that it is unknown whether the impact experienced by both groups was high or low relative to children without a significant malocclusion as there was no non-orthodontic control group.

There is, however, a dearth of research which has explored the psychosocial impact of AI during adolescence. One exception is a preliminary qualitative study [15] which found that adolescents felt self-conscious about their AI. They commented on how they were teased by peers, and that AI made some of them feel stigmatised and affected their friendships. These preliminary findings indicate that AI may be adversely affecting this age group.

It may also be the case that parents of adolescents with AI are affected and have specific support needs. Research has shown that parents who have a child with a health- related condition can also experience psychological distress. For example, Kunkel et al.[16] investigated parents who had a child with a facial hemangioma (a benign tumour). Compared with a control group, these parents demonstrated significantly lower levels of psychological wellbeing, with psychological distress being associated with severity of condition and medical complications. However, there is limited research exploring the effects of having a child with a dental disfigurement on the parents, including AI.

Online Support Groups

It may be the case that adolescents with AI and their parents have specific support needs that could be addressed via an Online Support Group (OSG). There are a number of advantages of such groups, for example they are not restricted by the temporal, geographical and spatial limitations typically associated with face-to-face groups, which can be

useful for those individuals who are living with a relatively rare condition. In addition, the anonymous nature of such groups can facilitate self-disclosure and help individuals in discussing sensitive issues more easily[17]. However, anonymity can also be a drawback, as less inhibited members could harass or disrupt the group[18]. Moreover, as many online support groups are un-moderated, opportunities exist for inaccurate, dangerous or mistaken medical information to appear[19] with misinterpretation a major risk[20].

The most common types of support participants in these groups report receiving are emotional and informational[21.] Although most groups are aimed at adults, there have been some specifically designed for children. For example, STARTBRIGHT World is a computer network that serves hospitalized children, providing interactive health education[22]. Children who participated in this network were found to be more knowledgeable about their health condition and had lower negative coping. This improvement in knowledge can lead the individual to a sense of empowerment which can have a positive impact on psychosocial well-being[18].

In summary, the limited research that has focused on the experience and perceptions of adolescents with AI, has highlighted that their condition may have adverse psychosocial effects. Parents of these adolescents may also experience challenges, though this population has not been explored in the research literature. Both adolescents and

parents may feel there is a role for OSGs in helping bring together individuals affected by AI and providing support through this forum.

The aims of this exploratory study are to explore the:

(1) experience and perceptions of AI from both the adolescent and their parent's perspective.

(2) views of AI adolescent patients and their parents as regards the usefulness of an online support group for patients/parents and;

(3) potential salient functions and features of such a resource (e.g., what do they consider the most important elements of an online support group?).

MATERIALS AND METHODS

FOCUS GROUPS

Focus groups were employed to help facilitate discussion between participants about their experience of AI and views on a possible online support group. In oral health research, focus groups have successfully been used to explore children's perspectives[23], thus we deemed them appropriate for use with the adolescents in this study, as well as their parents.

Sample

Participants were recruited from the Unit of Paediatric Dentistry at Eastman Dental Hospital (EDH) UCLH NHS Foundation Trust, using a purposive sampling strategy which is commonly used in qualitative research[24]. Eight participants were recruited for the study; four adolescents (patients) and their parent (either Mother or Father). To be included in the study, patients had to be English-speaking and between 11 – 16 years of age with a clinical diagnosis of AI for which they had received restorative care. The three males patients had severe, thin and pitted, hypoplastic AI, while the female patient had milder hypomature AI. None of the parents had AI.

Ethics

Ethical approval was obtained from Newcastle and North Tyneside Proportionate Review Ethics Committee as well as the Institute of Work, Health & Organisations in the University of Nottingham. All participants (patients and parents) were given information sheets to help them decide whether they wanted to take part in the study, and consent forms were signed prior to commencement of the study. All participants were informed that they could withdraw from the study at any time without it affecting their dental care, and were told that all data would remain anonymous and confidential.

Focus Group Schedule

A semi-structured focus group schedule was developed, drawing on open ended and broad questions to allow participants to express their

own experiences of AI and opinions of online support groups. The first author conducted both focus groups (with a co-author taking additional field notes) and these were both audiotaped. Half way through the focus groups, participants were shown examples of online support groups for acne and eczema. The different functions and features of these online groups were highlighted. Participants were given the opportunity to browse the online support groups if they wished to explore the features further.

Analysis

Thematic analysis is a method for identifying, analysing, and reporting themes within a data set. It minimally describes the dataset in rich detail, however it also often goes further than this and interprets various aspects of the research topic[25]. The data were analysed following Braun & Clarke's guidelines[26]. Firstly the focus groups were transcribed verbatim. Then the transcripts were read through thoroughly in order to identify major themes in the data. The next stage of analysis was labelling the themes so that they were easily identifiable, followed by an attempt to form a structure for the analysis. A major list of themes was then produced, with sub-themes emerging from each major theme.

Results

Details of all participants can be found in Table 1. All names have been changed to ensure participant anonymity. The focus group lasted for 37 minutes for the adolescents and 54 minutes for the parents.

TABLE 1 about here

There were three main themes that emerged from the data, they were 'Living with AI: Do I look bothered?', 'Need for the 'right' online environment' and 'Support needs: Information and beyond'. Each will be discussed in turn.

Living with AI: Do I look bothered?

What clearly emerged from the different focus groups was that responses differed in terms of how AI affects the adolescents. Often, clear contradictions took place. Firstly, when the adolescents were asked about how, if at all, AI affected them the general response was '*Not really'* or '*I don't mind having it'* with no discussion of adverse consequences. Grant specifically stated: '*yeah, I'm alright*'. However when parents were asked, Grant's mother Andrea commented:

'*He makes out it doesn't bother him but I think it does deep down. Definitely'* as well as *'it does bother him more than he lets on*.'

She went on further to explain a specific social situation where she noticed a change in Grant's behaviour:

'I go to parents evening and he'll be talking like this... [puts hands over mouth] *He doesn't want people to see, he does it without realising'.*

A similar experience was described by Debbie, the mother of Saul. She noticed that: *'when he's talking he closes his mouth and…err… he don't want to be smiling for pictures and covering his face. But it's funny because I'm like 'come on, smile properly you've got a very nice smile', but he goes 'no, I don't like it.''*

The adolescents spoke in some detail about how they do not like to speak to others about having AI. Frank comments that:

'Like you don't really want everyone to know. Cause you don't want them to get the wrong impression, like, you've got something really wrong with you.'

The adolescents made it clear that they still do not like to talk about having AI to friends. Grant says '*I just tell them I'm going to London to get my teeth done and that's it.*' After some prompting, Frank explored not speaking about AI to peers in more detail. He states that he does not speak about it purely down to the fact that it simply is not a conversation topic:

'*it's not really like brought up in conversation very often. You don't sit there and talk about teeth. Unless it's sort of like, I dunno, like, when with me its kinda been brought up like 'Oh you have really straight teeth'*

and I like joke saying 'yeah I go to London to get my teeth whitened,' in like a joke and make it fun.'

Need for the 'right' online environment

Overall, the adolescent participants were not entirely positive about an online support group. They were clear that they would only consider an online support group if it had certain salient features which would create the right online environment for them to get involved.

It was clearly identified throughout the focus groups that the adolescents did not want to engage with people they didn't know within online support groups. Aisha clearly states: *'I would only talk to people I know'.*

One of the parents, Debbie, also highlighted this as a potential drawback to discussion within a potential online support for AI:

'I think the kids will not share their problem with anyone. If they are both sharing at the same time and they know each other they will share… otherwise I don't think so… cause they are not going to add anyone randomly and share their problems'.

The parents themselves stated they would not want the online support group to be anonymous. One parent stated that from previous experience of a chat room she does not engage with strangers:

Andrea: *'I don't talk to anyone I don't know'*

A topic raised by some adolescents and their parents was whether there would be enough people to use the online support group and that a critical mass would be needed in order to keep it going. Sam in particular was very concerned and repeatedly brought this issue up in conversations. He first said:

'*If it's a large group it's ok, but if very small group then people may not use it*'.

Later on suggesting that: '*if there is a conversation going then, but you might not be able to maintain it. It might come to a halt or something'.*

Even though some parents voiced concerns that there would not be enough people to start off with to get the site up and running, Tom is optimistic and states: '*To start something off you need to sow the seed, that's my opinion. You need the seed and then it will grow and grow'.*

Within the adolescents' focus group, Aisha also voiced that she was concerned that '*people might not reply to your questions and ignore you'*.

There was frequent discussion surrounding the notion that an online support group must be appropriately moderated by a health professional, in this case a paediatric dentist who specialises in AI. As Tom says:

'*The last thing you want is someone going on who is a so-called expert and they're not, they're not an expert. Some mad dentist. [laughs] What you need to do is get it right'*. He repeats this idea later on in the focus group:

'Not having a person who doesn't have a clue of what they are talking about… as long as you stick to the criteria. A professional who are credible, so they do understand AI and they have been studying it'.

This was also spoken of within the adolescent focus group as well with Frank clearly commenting that:

'If it was just information from a dentist, it's nice to have someone who's experienced in it'.

Support needs: Information and beyond

It was clear that the adolescents felt an online support group, would be most useful if it was information based. The participants spoke clearly about what '*straight to the point'* information they would like to receive and again, wanting an immediate response was highlighted: Frank '*It would be better if…just asking any question and getting an immediate response'.*

Parents commented that they would benefit from general tips in helping to care for their child with AI. This theme became clear in the parents' focus group:

Sam: '*you could put all the information about what other treatment there is'.*

Tom: *'Local dentists for emergencies. Things like that'*

Tom: '*Dietary things is good as well. Cause you're not meant to eat sweets and things like that'.*

Sam: *'I think that cleaning the teeth, I'm not sure whether brushing is ok, or whether you are damaging it, yeah, giving advice'*.

Andrea spoke about a situation where she did not know the answer but if there was an online support group, she would have used it to get an answer:

'*My brother bought Grant this new toothbrush the other day cause I'm trying to encourage him to look after his teeth you know… and you know them pulsation one you can press the button, well I was thinking "will that damage his teeth?".*

Tom: 'see, you could have a section on there about toothbrushes'.

Although the parents discussed the potentially important role of information on an AI online support group, they went beyond this and spoke openly about needing other types of support.

Andrea states: '*It gets me down cause I wish he hadn't had it'*

Tom later references this and comments: *'Parent wise, as a support group of parents, as you [points at Andrea] say, it gets you [points at Debbie] down a bit, it gets you [Sam] down but you know…. A support group of parents, maybe we could ask each other questions'.*

On the day of the focus group, it was the first time both the adolescents and parents had met anyone else affected by AI. As well as the role of social support, parents discussed how they could be of practical help to one another including trying to arrange similar appointment times so they could share transport to London:

'*Well say we was booking an appointment say Frank's appointment was 11 o'clock, or in the afternoon, and you had one at 12 o'clock, I could ring you up and say 'I'm going up, we've got an appointment on the say day, do you want a lift?*' (Tom).

Discussion

The aim of the present study was to explore the experience and perceptions of AI from both the adolescent and their parent's perspective, as well as exploring what adolescents with AI and their parents thought about using an AI online support group; whether this would benefit them, and how.

There were three major themes that emerged across the adolescent and parent focus groups. One theme relates to the effects of living with AI. The findings were surprising. The adolescents reported that they are '*not bothered'* by having AI. This contrasts with previous research[9, 15,27] which found that patients may experience adverse psychosocial effects from having AI, and experience bullying and teasing from peers. There were clear contradictions however, between the reports of the adolescents and their parents. Parents described the way in which their

child covered their mouths in certain social situations, although one of the parents did indicate that their child may not even be aware of it. It may be that in certain social situations where possibly s/he is the focus of attention, a habitual behaviour has formed of trying to cover teeth in order to hide the affects of AI. This reflects previous findings[9] where the majority of young people with Developmental Defects of Enamel (DDE) reported feeling uncomfortable smiling freely. Moreover, Meaney et al[13] also found that adolescents with hypodontia altered their behaviour in order to hide their teeth as they felt they were not aesthetically attractive and so felt uneasy in social situations.

The fact that the adolescents expressed so keenly that they were 'not bothered' about having AI may well reflect their true feelings on the subject, indeed it may be that they are used to living with AI and do not see it has a 'big deal'. This certainly appears to be how they frame this when they discuss AI (if at all) with friends. Moreover, this fits with their assertions that an Online Support Group (OSG) for those with AI is not greatly needed for emotional support. The parents acknowledge that having a child with AI sometimes got them 'down'; their own feelings may possibly be projected on to the child leading them to conclude that '*deep down'* AI bothers the child too. However, there are also alternative explanations which should be highlighted. First, it should be noted that the responses may be a function of the study context. That is, it may be that the adolescents do not wish to reveal any concerns in front of

individuals that they have just met or because the first person to respond indicated that he was 'not bothered' so the rest followed his lead.

In terms of the online support group, it became clear that the adolescents did not highlight a specific need in terms of social support generally. This may well relate to their responses regarding 'not being bothered' by having AI, and experiencing no adverse effects in terms of bullying or teasing, and talking positively of their friendship groups. They also outlined how they wouldn't want to have discussions with people they didn't know and that they would not disclose information about themselves in the group. This is interesting, as previous research has shown that users of OSGs find the opportunity to have discussions with individuals who are not personally known to them an advantage and that anonymity is key to this[28]. Some of the participants (including parents) seemed to be comparing OSGs to chat rooms (or social networking sites – '*they are not going to add anyone randomly and share their problems*'), where talking to 'strangers' may be seen to be a dangerous activity which may well be the reasoning behind this. Although OSGs are different from chat rooms, these views would need to be considered if an AI OSG was to be taken forward.

Both groups of participants agreed that if a group was to be developed, then the key focus should be that of information. The adolescents emphasised the need to find information quickly and to also have speedy responses to questions or posts. A moderator who is a

dentist with expertise in AI was also seen as essential, to ensure that the right information was being posted. Ensuring information is correct on health-related internet sites is often cited as a concern. Indeed, opportunities exist for inaccurate, dangerous or mistaken medical information to appear on un-moderated sites[19]. As AI is a rare condition, input from an AI-specialist would be a valuable way to give advice and respond to queries to those affected, although the practical challenges of responding quickly would need to be considered.

One of the main findings that came from the focus groups was that parents need further support. As outlined previously, they were very positive about the information role that the OSG might potentially have, especially since they considered themselves responsible for their child's teeth in terms of day-to-day care, such as brushing and cleaning as well as long-term treatment plans. They also talked about how sometimes the fact that their child has AI gets them 'down'. This has not been found before in the literature, possibly because parents' views have only been explored in relation to their affected offspring. They spoke freely about how helpful it was to meet up with other parents of children with AI for the first time, and how they would like keep that communication going.

They acknowledged that a forum for discussion among parents would be valuable. In addition, the potential for practical or 'tangible' support came out specifically from the focus groups. Treatment for AI tends to be centred in main cities, where there are specialist AI paediatric

dentists. Therefore, many parents have to take a whole day to travel with their child from varying parts of England to attend appointments with financial and time implications. The parents discussed how they could keep in touch and co-ordinate appointments and share lifts. An online support group may be useful for these parents, and help provide a forum where other AI parents can access emotional, informational and practical support.

Limitations of the study

Recruitment was difficult due to the geographical spread of patients with AI attending EDH, and the rare nature of the condition. Even though the conversation flowed well and participants all contributed, it would be interesting to see if there was another focus group using the same question structure, if the findings would be the same. In addition, participants in the focus groups volunteered to take part in the study, thus they may differ from patients with AI who did not choose to participate, in terms of levels of social support needed and views on an online support group. Also, it is important to note that there are many clinical forms of AI, so we cannot assume that these views represent all adolescents with AI. Therefore, overall, as qualitative research is context specific, our findings cannot be generalised to all adolescents with AI.

Conclusion

In conclusion, this exploratory study found that these adolescents did not appear to experience adverse effects of having AI, although their

parents were not of the same opinion. An online support group would need to be primarily information based and moderated by an AI specialist. Parents would benefit from additional features which facilitate support beyond that of information, such as emotional and tangible support, perhaps in the form of a parent discussion forum. It is suggested that at first, users of such a group would be patients from Eastman Dental Hospital, as participants stated the importance of privacy and communicating with people who have been clinically diagnosed with AI. Adolescents and parents should continue to be consulted in any development of an AI OSG, and any such group should be monitored and evaluated.

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